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**Diagnosis and Management of Iliac Artery Endofibrosis:**

**Results of a Delphi Consensus Study**

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**(INnternational Study group for Identification and Treatment of Endofibrosis)**

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## **Abstract**

## **Introduction**

Iliac endofibrosis is a rare condition that may result in a reduction of blood flow to the lower extremity in young otherwise healthy individuals. The data to inform everyday clinical management are weak and therefore a Delphi consensus methodology was used to explore areas of consensus and disagreement concerning the diagnosis and management of patients with suspected iliac endofibrosis.

## **Methods**

A three-round Delphi questionnaire approach was used among vascular surgeons, sports physicians, sports scientists, radiologists and clinical vascular scientists with experience of treating this condition to explore diagnosis and clinical management issues for patients with suspected iliac artery endofibrosis. Analysis is based on 18 responses to round 2 and 14 responses to round 3 with agreement reported when 70% of respondents were in agreement.

## **Results**

Initially there was agreement on the typical symptoms at presentation and the need for an exercise test in the diagnosis. Round 3 clarified that duplex ultrasound was a useful tool in the diagnosis of endofibrosis. There was consensus on the most appropriate type of surgery (endarterectomy and vein patch) and that endovascular interventions were inadvisable. The final round helped to inform aspects of the natural history and post-operative surveillance. Progression of the disease was likely with continued exercise but cessation may prevent progression. Surveillance after surgery is generally recommended yearly with at least a clinical assessment.

## **Conclusions**

There is broad agreement about the presenting symptoms and the investigations required to confirm (or exclude) the diagnosis of iliac endofibrosis. There was consensus on the surgical approach to repair. Disagreement existed about the specific diagnostic criteria that should be applied during non-invasive testing and about post-operative care and resumption of exercise.

## **Introduction**

The onset of exercise-induced leg pain in young, otherwise healthy individuals can often lead to diagnostic difficulty, particularly when those affected are high-performance athletes.

Over recent years it has become apparent that these symptoms could be caused by non-atherosclerotic lesions. Among these are endofibrosis of the arteries specifically of the external iliac artery and, perhaps lengthening and kinking of the external iliac artery.<sup>1 2</sup> There is an increasing awareness of iliac endofibrosis, particularly among elite sports people (especially cyclists) consequent to a number of high profile cases. Yet iliac endofibrosis is a condition that is infrequently managed by vascular specialists and often presents to non-specialists and allied healthcare professionals. Consequently patients with the condition may have a delayed diagnosis or be incorrectly managed.<sup>3</sup>

A recent systematic review of the literature has confirmed there is a paucity of robust data to inform the diagnosis and management of patients with iliac endofibrosis.<sup>4</sup> At present there are no guidelines or standardised pathways of care to ensure otherwise healthy individuals receive consistent advice and appropriate management of their condition.

This study reports the results of a Delphi consensus among members of INSITE (INternational Study group for Identification and Treatment of Endofibrosis). INSITE is an international inter-disciplinary group of healthcare professionals with a common interest in the management of people with iliac endofibrosis and comprises specialists from sports medicine, sports science, vascular surgery, radiology and medical imaging. The objective of this study was to identify areas of consensus in diagnosis and management of iliac endofibrosis and to provide a starting point for the future development of guidelines for best practice.

## **Methods**

The Delphi method is a structured communication technique developed as a systematic, interactive forecasting method. It relies on experts answering questionnaires in two or more rounds. After each round, a facilitator provides an anonymous summary of the experts' forecasts from the previous round as well as the reasons they provided for their judgments. The experts are therefore encouraged to revise their earlier answers in light of the replies of other members of the panel. During this process it is anticipated that the range of answers will decrease and the group will converge towards a 'correct' answer or 'consensus'.

A three-round Delphi questionnaire approach was used<sup>5</sup>. The first round of the questionnaire was generated from discussion among experts: vascular surgery (n=3), sports medicine (n=2), radiology (n=1) and clinical vascular scientist (n=1)) and comprised eight broad areas of iliac endofibrosis (with or without iliac kinking) management. Within these eight areas a number of questions with stems were generated (Table 1). Some specialists did not complete specific aspects of the questionnaire that were outside of their area of expertise (for example some of the technical aspects of surgery were not necessarily completed by non-surgeons). The second round questionnaire was distributed to the participants who were all members of INSITE in June 2015. They were invited to complete it and provide any specific comments they perceived to be necessary. The results were collated from the second round. Consensus was achieved when 70% of respondents were in agreement. A third and final round of questionnaires was sent out to the same group of specialists in September 2015. The questionnaire comprised the same questions. Each specialist had access to the second round scores for the whole group as well as their own personal score from the second round, allowing them to compare their views to those of the group and enabling improved evidence for consensus items from the second round. Respondents were invited to re-score the questions in light of the data from the second first round using a Likert scale.<sup>6</sup> The 4-point Likert scale (strongly agree to strongly disagree 1-4) allowed strongly positive responses to be recorded as 1 and strongly negative responses as 4, but has no neutral position and forces a choice. In cases where the respondents' new scores differed from the group score, they were invited to comment to ensure the question was correctly interpreted and expose common fallacies.

## **Results**

In the second round of the Delphi consensus, responses were received from 18 specialists. The overall responses to key questions are shown in table 1. The third round of the Delphi received responses from 14 specialists and led to consensus on a number of issues which are underlined in table 1, as well as a number of useful comments also reported in table 1.

### *Diagnosis*

In the second round there was consensus that leg weakness, thigh pain and resolution of symptoms within 5 minutes of cessation of exercise were all hallmarks of iliac endofibrosis.

After the third round, clinical assessment and duplex ultrasound were thought to be important in the assessment and diagnosis of iliac endofibrosis. The respondents felt that an exercise test (measuring pre- and post-exercise ankle pressure / ABPI) was the most appropriate way to confirm (or exclude) iliac endofibrosis. No specific exercise test is required but there was consensus that the patients should exercise until they reproduced their symptoms. Initially there was no consensus on how rapidly the subject's ankle pressure measurements should be measured after cessation of exercise. However after round 3 most felt that it was important to measure pressure as quickly as possible (within 1 minute of exercise cessation). There was no consensus on what absolute pressure drop constitutes a positive test for iliac endofibrosis, although a pressure drop between both legs in an individual patient with unilateral symptoms of between 21-40mmHg was thought to imply a positive test. There was no consensus reached about the absolute level of ABPI at cessation of exercise. There was consensus that measurement of ankle pressures and ABPI was best undertaken with the patient in the supine position. No information was sought on whether manual or automatic blood pressure cuffs were used.

### *Natural history*

In the second Delphi round, there was consensus that the number of hours of cycling was associated with the development of endofibrosis. There was no consensus on whether arterial kinking alone is sufficient to cause flow limitation, or on the effect of continuing or stopping exercise. After the third round, however, there was consensus that arterial kinking in isolation was not responsible for limiting blood flow during exercise and that cessation of exercise prevents the progression of the disease (rather than causing regression) whilst continuing to exercise leads to disease progression.

### *Treatment*

In the second Delphi round there was immediate consensus that medical therapies are ineffective in the management of endofibrosis, that surgery should be offered to anyone with severely impacted quality of life and that endovascular interventions have no role in patients with endofibrosis. There was, however, wide variation in views on the most appropriate advice to give to a person with a new diagnosis of iliac endofibrosis.

After the third round there was greater consensus, with agreement that clinicians must decide on an individual patient basis whether cessation of cycling should be considered as a first line

169 of treatment. It was also agreed that changing cycling position may help relieve some  
170 symptoms.

### 171 *Surgery*

172 Shortening of the external iliac artery was thought to be ineffective as a sole strategy for the  
173 treatment of endofibrosis. There was consensus that the extent of surgery should be guided by  
174 intra-operative findings, however, after the third round of questions pre-operative imaging  
175 was thought to be a helpful adjunct in guiding the extent of the procedure. There was  
176 consensus that an endarterectomy should be used where possible but that inguinal ligament  
177 release, prosthetic patches and bypass should not routinely be used in surgery. In patients  
178 with bilateral disease surgery should be performed one side at a time. The majority of  
179 specialists suggested that bypass surgery is generally preferable in patients who present with  
180 an occluded external iliac artery but no consensus was reached.

### 181 *Post-operative management*

182 There was no consensus on the duration of cessation of exercise / sport ('stand-down')  
183 following surgery, although there was consensus that some period of stand down was  
184 required following surgery (54% agreed that a 6-8 week period was most appropriate).  
185 Agreement was reached that all patients should receive aspirin following surgery but the  
186 duration of treatment remains unclear. There was consensus that surgeons should ideally  
187 perform an exercise testing to confirm that the treatment had been successful but some  
188 commented that merely the absence of symptoms was sufficient to suggest that the treatment  
189 had been successful.

### 190 *Long-term follow-up*

191 After the first round there was consensus that all patients should receive regular surveillance.  
192 There was no consensus on what form of surveillance patients should receive, however after  
193 the second round of questions there was general consensus that this should at least comprise  
194 clinical assessment on a yearly basis. Most (64%) of the respondents suggested that (non-  
195 invasive) imaging post-operatively was helpful as it may help identify pseudoaneurysms or  
196 other potential complications that might otherwise be asymptomatic.

### 197 *Management of post-op complications*

In terms of the management of specific local surgical complications there was agreement that a stenosis should only be considered for treatment if symptomatic and the optimal way to manage these stenoses was surgically (91%) and that patients should be advised to stop exercising (91%). There was a clear indication that intervention for widening or aneurysmal disease subsequent to patch angioplasty was indicated at >3.0 cm and for focal pseudoaneurysms and that these should normally be repaired surgically.

### *Screening*

There was no consensus on the need for screening any groups of asymptomatic individuals for iliac endofibrosis but 57% suggested that screening in professional cyclists may be justified (an exercise test would be the optimal screening method). If endofibrosis is detected surgical intervention should only be considered if the patients develop symptoms.

### **Discussion**

There are no guidelines or standard care pathways for the assessment and management of patients with suspected iliac endofibrosis. Iliac endofibrosis is a rare condition and consequently has few published data on which to draw firm conclusions about best practice. Until these data are available it is quite possible that patients may suffer unnecessarily from delayed diagnosis and inappropriate management. In the absence of randomised trials or other controlled studies the use of Delphi consensus methodology to develop consensus among specialists is a reasonable alternative to inform clinical practice.

There was broad agreement that patients usually present with a symptom complex of leg weakness and thigh pain, typically resolving within 5 minutes of exercise cessation. Non-invasive testing and imaging are usually the first line in the diagnosis, in line with published data.<sup>7</sup> Although an exercise test and duplex ultrasound are helpful in the diagnosis of the condition the absolute values of pressure drop in the lower limb represent a significant area of controversy with a drop of between 21-40mmHg being considered diagnostic. However, this controversy may simply reflect that endofibrosis is a progressive disease. In the early stages the symptoms are exclusively at high intensity exercise and the pressure drop is low, in more advanced disease the symptoms commence during lower exercise intensity and are associated with a larger pressure drop. Similarly the duplex criteria for the diagnosis of endofibrosis are poorly defined.<sup>8, 9, 10</sup>



A consensus that exercise tests should be standardised was reached, which is an important step forward, and a description of this technique was recently published.<sup>11</sup> Patients should be exercised until they develop symptoms and the pressures (ankle and ABPI) should be measured if possible within one minute of exercise cessation with the patient in a supine position (and for this some units have adopted automatic systems to measure pressures simultaneously and rapidly in both lower limbs). Some institutions, that consider kinking as a potential important cause of flow limitation, test the ankle pressures with the patient on a cycle ergometer in an upright position (with a correction being applied for vertical height difference) and with hips flexed<sup>2</sup>.

Management of iliac endofibrosis should always include a careful discussion with the patient and cessation of exercise should always be considered by the athlete before surgery is undertaken. Comments provided by the Delphi participants suggested these discussions are frequently difficult as young fit sports people rarely wish to stop exercising. However, this dialogue is important given the paucity of robust surgical outcome data either in the short and more particularly the long-term.<sup>4</sup> Intervention is not without the potential for serious complications and surgery should only generally be recommended for those with symptoms causing a significant impairment of their quality of life (and after risks of the procedure have been fully explained).

Original data from the Netherlands had suggested that kinking of the iliac arteries was associated with flow limitation.<sup>2</sup> However, surgical outcome data from the same unit and others suggested shortening of the iliac arteries in isolation (without endarterectomy and patching) should only be performed in very selected cases where there is no stenotic disease as otherwise it may lead to sub-optimal outcomes.<sup>13</sup> The Delphi consensus corroborated these findings with most experts suggesting that kinking alone is rarely a cause of significant symptoms and surgically removing a kink will be largely ineffective if performed as a sole manoeuvre and should be avoided. Though preoperative imaging may provide some guidance on the length of the endofibrotic segment, it was generally agreed that the extent of the endarterectomy should be determined by operative findings. However, comments from some Delphi participants suggested that intra-operative angioscopy may also be useful in guiding the extent of the procedure.

In conclusion, although there was diverse opinion in some areas, consensus was reached on a number of key aspects of iliac endofibrosis. Consensus was reached on the typical presenting

symptoms and the non-invasive testing required to confirm the diagnosis. Most experts believe that cessation of exercise prevents the progression of disease and should be recommended to most patients as a first line management strategy. The type of operation employed should involve an endarterectomy and vein patch repair of the external iliac artery, endovascular therapy and treating kinking alone should be avoided.

The findings of this Delphi consensus study should be used to inform patients with suspected iliac endofibrosis and all healthcare professionals who may be involved in their care. This study has highlighted areas that require further research. As a first step a registry (planned) of patients undergoing surgery will help capture important data on outcomes that may influence clinical practice. Furthermore, guidelines to standardise care and improve management and outcomes and guide research in the areas of need are warranted.

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